FERTILITY AND STERILITY®
VOL. 70, NO. 4, OCTOBER 1998

Published by Elsevier Science Inc.
Printed on acid-free paper in U.S.A.

Intermittent azoospermia associated with epididymal sarcoidosis

David A. Svetec, M.D., Robert L. Waguespack, M.D., and Edmund S. Sabanegh, Jr., M.D.

Wilford Hall Medical Center, San Antonio, Texas

Objective: To report an unusual case of intermittent azoospermia associated with epididymal sarcoidosis.

Design: Retrospective case analysis.

Setting: Wilford Hall Medical Center.

Patient(s): A 36-year-old male with secondary infertility and epididymal sarcoidosis.

Intervention(s): None.

Main Outcome Measures(s): An analysis of sperm count in relation to steroid courses.

Results(s): Epididymalgia, and to a lesser extent, sperm counts were noted to fluctuate temporally around steroid courses given for pulmonary flares of sarcoidosis. Epididymal sarcoidosis can be associated with intermittent azoospermia. Presumably, epididymal granulomas undergo exacerbations and remissions and cause intermittent ductal obstruction.

Conclusions(s): Because of the unpredictable effect of sarcoidosis on the male genital tract, all patients interested in paternity should obtain a semen analysis at the time of disease diagnosis. If oligospermia is noted or if there is clinical evidence of epididymal involvement, the patient should be offered sperm banking for possible future assisted reproductive techniques. (Fertil Steril® 1998;70:777–9. ©1998 by American Society for Reproductive Medicine.)

Key Words: Infertility, sarcoid, epididymis, azoospermia

Sarcoidosis is an idiopathic, multisystem disease characterized by epithelioid, noncaseating granulomas. The disease is estimated to affect 1 in 10,000 people in the United States resulting in roughly 22,500 cases annually. To establish sarcoidosis as a diagnosis, a patient must display a clinically and radiographically compatible picture and demonstrate noncaseating epithelioid cell granulomas in biopsied tissue, after other causes of granulomatous infiltration are excluded. Sarcoidosis most commonly presents with intrathoracic manifestations (84%) but can involve virtually any organ system. Open lung biopsy has been the standard diagnostic approach, but transbronchial biopsies, thorascopic biopsies, mediastinoscopy, scalene node biopsy, and liver and muscle biopsies have all been used.

Involvement of the genital system is relatively rare, with a reported incidence of 4%–4.5% in postmortem examinations and 0.5% clinically (1–3). Genitourinary manifestations of sarcoidosis include renal involvement with nephrocalcinosis, uremia, and genital lesions including granulomas of the epididymis, testis, and vas deferens as well as cutaneous genital lesions. We report a case of epididymal sarcoid with resulting secondary infertility. Salient clinical features and fertility management options are reviewed.

Received January 21, 1998; revised and accepted May 15, 1998.

This manuscript was prepared by officers of the United States Government as part of their official duties. No separate financial support was utilized in the preparation of this manuscript.

Reprint requests: Edmund S. Sabanegh, Jr., M.D., Department of Urology, Wilford Hall Medical Center, 2200 Bergquist Drive, Suite 1, Lackland AFB, Texas 78236-5300 (FAX: 210-292-7199).

0015-0282/98/$19.00
PII S0015-0282(98)00271-4

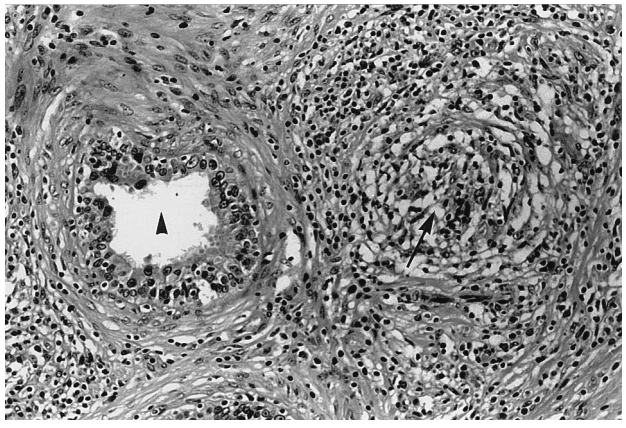
CASE REPORT

A 36-year-old male was referred for evaluation of secondary infertility. The patient had fathered three children born in 1978, 1980, and 1982 with a previous partner. He was diagnosed with sarcoidosis by an inguinal lymph node biopsy in 1983 after presenting with joint pain and diffuse lymphadenopathy. The patient had a normal semen analysis in 1986 as part of an infertility evaluation. He subsequently developed bilateral painful epididymal nodules.

Scrotal ultrasound (US) examination revealed a 3.5-cm nodule in the globus major of the left epididymis as well as diffuse bilateral epididymal enlargement. He underwent a left partial epididymectomy in 1991 that showed

FIGURE 1

Photomicrograph of partial epididymectomy specimen showing epididymal tubule (*arrowhead*) and adjacent noncaseating granuloma (*arrow*). Original magnification, ×40.



granulomatous epididymitis consistent with sarcoidosis (Fig. 1).

Over a 10-year period starting in 1986, the patient had numerous semen analyses with counts varying from 0 to 45×10^6 sperm/mL (Fig. 2). On three occasions, he had normal volume azoospermia with follow-up analyses which revealed normal counts or mild oligospermia. Additional urologic evaluation included a post-ejaculate urinalysis which showed no sperm. Serum FSH and LH levels were normal.

Repeat scrotal US examination showed right epididymal enlargement with a small left hydrocele and normal testes in September 1995 (Fig. 3). During this period, he received multiple short courses (2–4 weeks) of oral steroids for pulmonary sarcoid flares. After each steroid treatment, he noted marked improvement in his epididymalgia with resolution of the epididymal enlargement within several weeks, lasting up to 3 months at a time.

A recent course of steroids resulted in no decrease in his epididymalgia, and a semen analysis showed persistent azoospermia. At the present time, he is being considered for epididymal aspiration with intracytoplasmic sperm injection (ICSI).

DISCUSSION

Although rare, sarcoid involvement of the genital tract has been documented in the testis, epididymis, prostate, penis, and scrotal skin. Testicular and epididymal sarcoidosis must be differentiated from other causes of granulomatous diseases that affect these tissues, including tuberculosis, syphilis, sperm granuloma, filariasis, lymphogranuloma venereum, granuloma inguinale, atypical tuberculosis, blastomycosis, coccidioidomycosis, actinomycosis, schistosomiasis, and Wegner's granulomatosis (4).

Granuloma of the epididymis is the most common manifestation of sarcoid involvement of the genital tract (5). Epididymal involvement is usually unilateral, nodular, and painless; however, bilateral lesions and patients who experience either acute or recurrent epididymitis have been described (6). Sarcoid involvement of the epididymis usually shows a periductal distribution and can lead to azoospermia (4). It can also progress to irreversible fibrosis and severe organ dysfunction (6).

Spontaneous remission of epididymal involvement can occur. The role of corticosteroids in treating epididymal involvement is unclear. Some reports document resolution of granulomas after corticosteroid treatment, whereas in other reports, epididymal involvement develops or progresses while patients are on steroids (7). Symptomatic epididymal

FIGURE 2

Relationship of semen analysis and steroid courses.

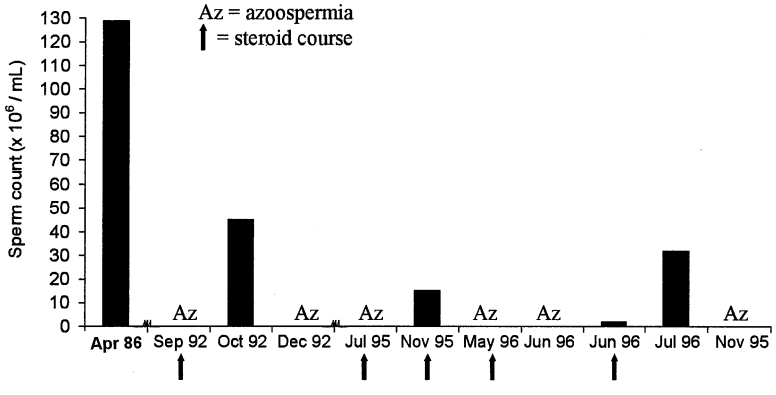


FIGURE 3

Scrotal ultrasonogram displaying E, epididymal nodule, and testis (*arrowhead*).

sarcoidosis may require excisional biopsy for cessation of pain (6).

This report represents the first description of intermittent azoospermia associated with sarcoid involvement of the epididymis. Other causes of intermittent azoospermia have not been identified. Although obstruction was not proven formally by testis biopsy or vasography, it is unlikely that variations in testis sperm production can explain these dramatic serial changes in semen quality. This case illustrates that a patient of normal fertility at the onset of the sarcoidosis diagnosis can progress to azoospermia. Although some temporal relationship is suggested between short-course steroid administration and improvement in the semen analysis, this phenomenon may alternatively represent the ability of sarcoid granulomas of the epididymis and the resultant ductal obstruction to undergo spontaneous exacerbations and remissions.

Because of the unpredictable effect of sarcoidosis on the male genital tract, it is recommended that all patients interested in paternity obtain a semen analysis at the time of disease diagnosis. If oligospermia is noted or if there is clinical evidence of epididymal involvement, the patient should be offered the use of sperm banking for possible future assisted reproductive techniques. Patients who present with azoospermia should have serial semen analyses to detect temporary resolution of the epididymal obstruction and to allow for the use of sperm banking during those intervals. Steroid therapy may assist with the transient restoration of genital tract patency, although this treatment should be studied in a prospective manner.

References

1. Ricker W, Clarke M. Sarcoidosis: a clinicopathologic review of 300 cases including 22 autopsies. Am J Clin Pathol 1949;19:725.
2. Engle R Jr. Sarcoid and sarcoid-like granulomas: a study of 22 autopsies. Am J Clin Pathol 1953;29:53.
3. Scadding J. Sarcoidosis. London: Eyre and Spottiswoode, 1967:417.
4. Rudin L, Megalli M, Mesa-Tejada R. Genital sarcoidosis. Urology 1974;3:750–754.
5. Opal S, Pittman D, Hofeldt F. Testicular sarcoidosis. Am J Med 1979; 67:147–150.
6. Ryan D, Lesser B, Crumley L, Cartwright H, Peron S, Haas G, Bower G. Epididymal sarcoidosis. J Urol 1993;149:134–136.
7. Winnacher J, Becker K, Katz S, Matthews M. Recurrent epididymitis in sarcoidosis: report of a patient treated with corticosteroids. Ann Intern Med 1967;66:743.